Psychic Stress as a Trigger of the Spontaneous Development and Rupture of an Aneurysm?

A Case Report

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Summary

We present angiographical and neuropathological data of a 51-year-old woman, who primarily suffered from a recurrent glomus temporale tumour on the left side and who finally died after a spontaneous rupture of a distal basilar aneurysm which developed during angiography. As a second important finding the existence of a vasospasm at the time of the rupture is also confirmed.

Case report

A 51-year-old woman was sent to our institute in order to embolize preoperatively a large recurrent chemodectoma of the left temporal bone and upper cervical spine. The patient had first undergone surgery 16 years before.

A documented huge recurrent tumour had been operated (after preoperative particle embolisation as well) two years before, knowing that a tumour rest stayed at the level of C1 / 2. Because of the primary tumour and the multiple operations, the neurological deficit was extensive: besides an amaurosis of the left eye and a rest vision of the right eye she presented a paresis of the left cranial nerves III - XII. The last MRI three months before had shown an extensive tumour invasion of C1 and C2 as well as the whole left skull base but a normal configuration of the upper brainstem. Current cerebral four vessel angiography disclosed the known (from earlier controls) occlusion of the left common carotid artery (CCA) about 2 cm distal to the aortic arch because of the occlusion of an external DACRON-bypass from the left common carotid artery (CCA) to the cavernous segment, set at the time of the last operation.

Consequently the ipsilateral internal carotid artery (ICA) did not opacify but a complete supply of the left hemisphere by the right ICA and the basilar artery via posterior communicating artery (Pcom) was seen. Because of the bypass, the left external carotid artery (ECA) was occluded, but part of the ipsilateral external vascularisation of the recurrent tumour was recanalised by opposite external arteries, not accessible by endovascular catheterisation.

The sources of most of the angiographically reachable tumour vessels were mostly capillary-sized ipsi- and contralateral muscular branches of the vertebral artery (VA) (figures 1, 2A, 2B).

The strategy of the interventional operation included a particle embolisation of the tumour-feeders under temporary balloon-occlusion of the left VA, proximal to the branching of the posterior inferior cerebellar artery (PICA). In order to control the consciousness of the patient, she did not receive general anaesthesia but the operation was done under sedation.

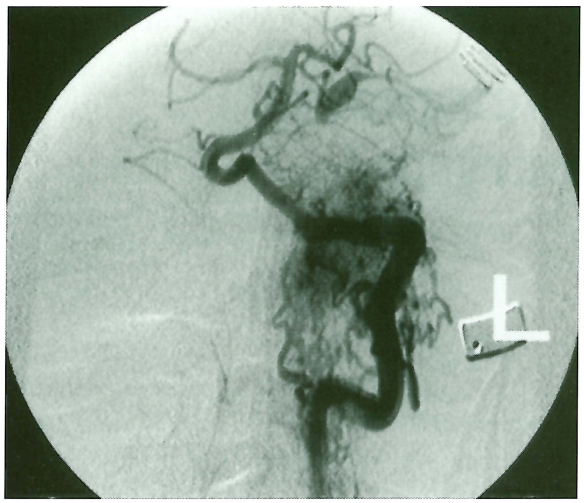

Figure 1 DSA of the left vertebral artery, demonstrating the tumour supply by muscular branches of the extracranial part of the VA. The branching of the right superior cerebellar artery is covered by the right P1-segment in an asymmetric configuration of the top of the basilar. Notice the anastomotic supply of the left supratentorial circulation via the left Pcom.

Results

Diagnostic angiography did not show any suspect change of the calibre of the distal BA, which presented a dilated left P1-segment and curved Pcom as well (figures 1, 2A, 2B). The interventional procedure started with particle embolisation of the feeders of the right VA, its calibre and opacification being smaller than that of the left VA.

Control angiography showed the occlusion of the feeders, but no remarkable changes of the distal basilar artery.

The decreased opacification of the distal BA is due to the small calibre of the right VA and TOWN projection, where the right P1-segment overlapped the proximal right superior cerebellar artery (figure 2C). The distal part of the basilar artery was seen unaffected in the lateral view (figure 2D), similar to the first projection, where especially the proximal part of the superior cerebellar arteries and the P1-segment of the left PCA (with Pcom) were delineated without any suspicion of an aneurysm. During catheterisation of the left vertebral artery to place the guiding catheter and balloon, the patient suddenly lost consciousness with a deep groan. She developed an asystole and increase in blood pressure (230/120 RR) followed by an extensive extrasystole.

After immediate fiberoptic intubation the angiography of the left VA (about 30 minutes later) only showed an opacification of the tumour, but no distal VA nor basilar artery (figure 3).

Injection of the right VA disclosed an extensive extravasation of contrast material at the right distal part of the BA, at the level of the branching of the superior cerebellar artery with a narrowing of the distal part of the BA and a retrograde filling of the narrowed intracranial part of the left VA (figure 4).

Immediate cranial CT showed an extensive subarachnoid and intraventricular haemorrhage, mixed with contrast material as well as an acute hydrocephalus (figure 5).

The whole intervention was performed under sedatism, i.e. a stable sedation with the benzodiazapine METHIMAZOL (Dormicum®). Contrary to the previous procedures and despite the fact that the patient was informed about the procedure and knew the physician, she was restless, anxious and impatient, always asking if everything went well and how long the procedure would last. Half an hour before the rupture, the anaesthesiologist recorded an "emotional imbalance", which was treated with another milligram of Dormicum R. Blood pressure at that time was about 190/100 mm Hg, whereas it was 140/80 at the beginning of the procedure.

The patient died two days later and was brought to autopsy, to establish the reason for the unexpected course of the intervention.

At autopsy a special preparation technique was used to investigate the pathologic alterations in the cranio-spinal region: the posterior cranial fossa with the cerebellum and the lower brain stem was removed in connection with the cervical spine and spinal cord. After formalin fixation all anatomical details of the cranio-spinal junction were prepared carefully: central nervous system, nerves and blood vessels as well as meningeal and osseous structures and especially the tumour in the skull base. An extensive intracranial and intraspinal haemorrhage was confirmed (figure 6).

After removal of the blood clots a freshly ruptured distal basilar aneurysm was demonstrated, without a circumferential wall or any thrombotic material (figure 7).

Macro- and microscopically the specimen did not look different from other ruptured berry

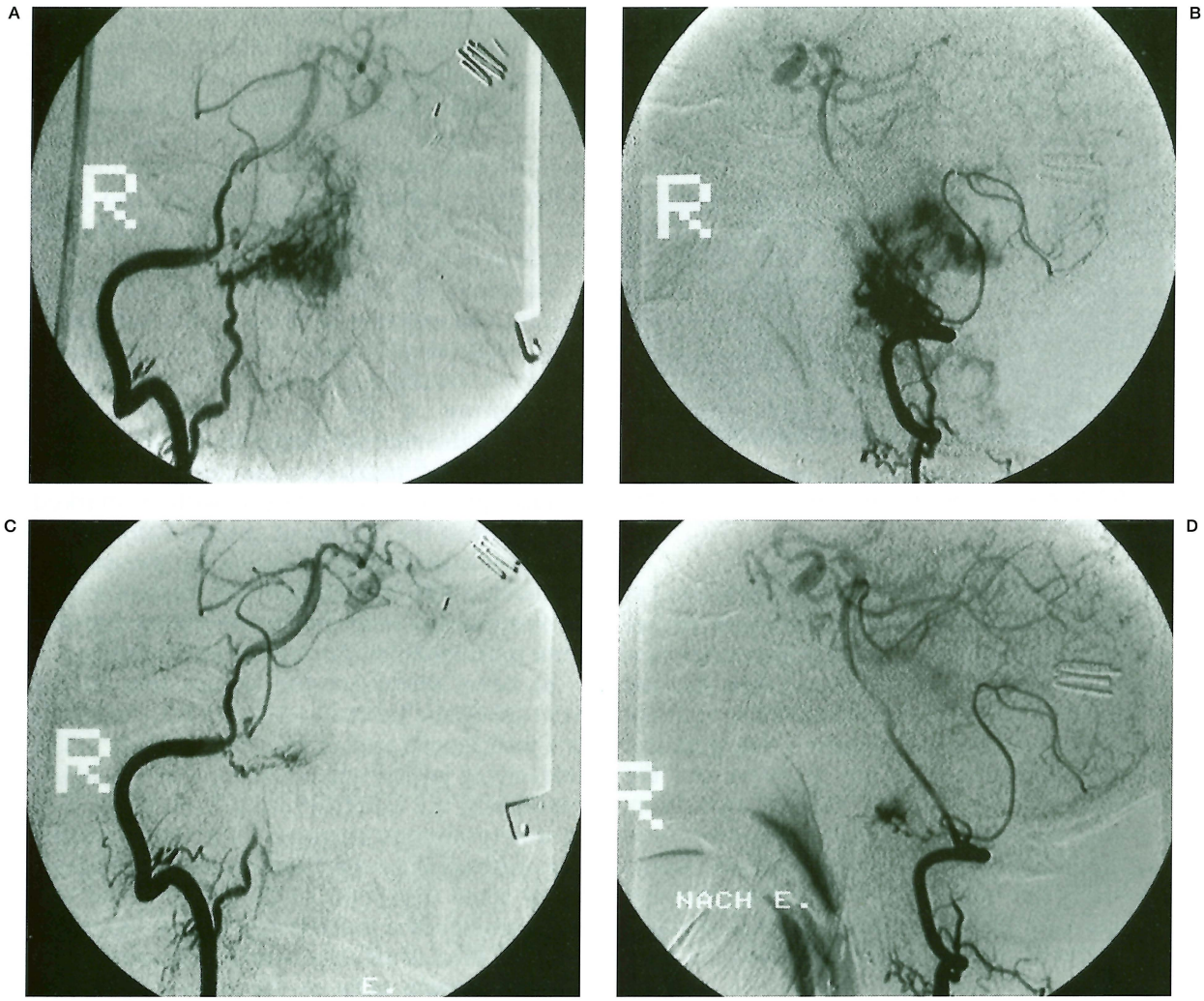

Figure 2 A) Preinterventional DSA of the right VA (ap view) shows tumour opacification by a big muscular branch. Because of a smaller diameter of the right VA, the corresponding opacification of the distal BA is less. B) Lateral view at the same time. C) Ap-view after particle embolisation with opacification of the proximal muscular artery. Although the overall opacification of the distal BA is better than in (A), only infratentorial arteries are seen. One may suspect a slight dilatation of the proximal superior cerebellar artery, not clearly differentiated from an overprojection of the ipsilateral P1-segment. D) The lateral view of the right VA after embolisation does not show any objective dilatation of the curved basilar tip nor of the right superior cerebellar artery suspect for an aneurysm (better contrast than preinterventional).

aneurysms, where an abrupt tear of the internal elastic lamina is seen (figure 8), besides the fact that the "wall" of the aneurysm consisted only of fibrinoid material and remnants of the adjacent leptomeninges. But even in parts of the vertebrobasilar vessels remote from the ruptured aneurysm different changes in the internal elastic lamina were seen. There were scars in the internal elastic lamina, representing different stages of older lesions of the blood vessel wall with focal haemorrhages in the adventitial layer (figures 9, 10).

Discussion

To our knowledge this is the first time that the obvious development as well as the rupture of an aneurysm in about one hour has been documented angiographically. New concepts in the development of cerebral vessel pathology, being aneurysm and / or arteriovenous or venous malformations, favor a trigger for the pathological changes, being intrinsic, psychic or of unknown origin[1]. One may imagine the extent of the psychic stress of this patient, given the acute per-

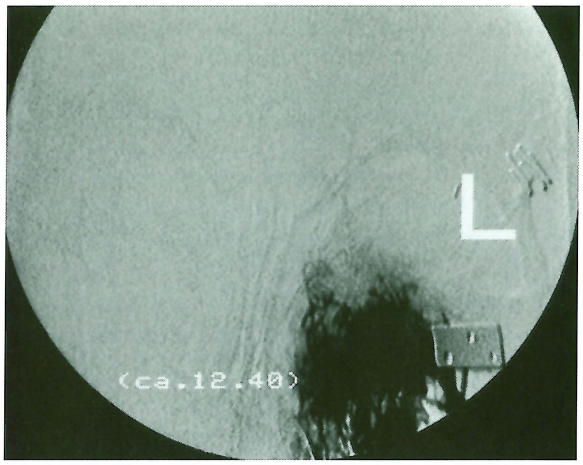

Figure 3 After the clinical event (see text) the injection of the left VA only visualises the extensive tumour supply, but no distal territory.

sonal history of the patient, which was related to us after the event: although she knew of her recurrent tumour, she did not really suffer new symptoms and did not feel worse than she had in the last 14 years. She was tired of the multiple operations and hospitalisations of the past and was only persuaded by her husband and the neurosurgeon to undergo another operation. She told her husband that she was convinced she would not return home, but that she would die during her hospital stay. Therefore, the restlessness and obvious anxiety which dominated the intervention even under sedation, can surely be interpreted as her fear of dying at that time. In preoperative and intraoperative management anaesthesiologists consider the fact that emotions like fear, anxiety and rage stimulate the output of adrenaline and noradrenaline in a different manner. Rage will activate the distribution of adrenaline, whereas fear and anxiety will activate noradrenaline production, resulting in a rise in blood pressure [2,3,4].

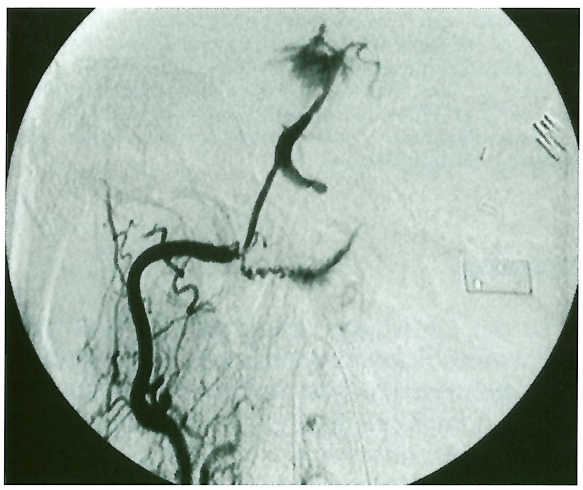

Figure 4 The injection of the right VA discloses extravasation of contrast material at the site of the rupture at the distal BA and retrograde filling of the left V4-segment. Note the narrowing of the distal part of the BA and the intracranial part of the left VA as well.

Therefore the maximum of psychic stress our patient was exposed to resulted in a vegetative state with slowly ascending blood pressure, which on the other hand was thought to lead to the rupture of one of the already present defects of the internal elastic lamina of the basilar artery. Metimazol R is the best substance to reduce preoperative anxiety with a marked reduction in the output of catecholamines, the reason it is used daily [5]. Despite the patient was medicated with Metimazol®, her existential fear of dying at any time during the planned procedure predominated during the intervention.

In berry aneurysm the abrupt tear of the internal elastic lamina and rupture of the media is a well known and often described fact [6,7]. The underlying pathologic alterations of the blood vessel wall are quite different from those seen in atherosclerotic changes. There are probably two pathogenetic mechanisms finally resulting in an acute dilatation of the blood vessel or - even worse -in a sudden rupture of the vessel wall [8,9,10]. First of all degenerative changes in the smooth muscle cells in the tunica media (in small arteries of muscular type) probably precede the acute rupture. Many chemical substances are known tp cause toxic damage of smooth muscle cells, such as adrenaline, noradrenaline, nicotine and many other sympathicomimetic drugs. Degenerative changes to the connective tissue of the blood vessel wall could also contribute to the damaging process, e.g. in lathyrism or in chronic degenerative disease of the connective tissue [11].

The second process is obviously the rupture of elastic membranes, especially of the internal elastic membrane. This event could be interpreted as a consequence of the insufficiency of the arterial muscle wall in resisting to an acute elevation of blood pressure induced by stress or sympathicomimetic drugs.

Beside the fact that in this special case angiographic projections of the distal BA were suboptimal (since the focus was the site of the

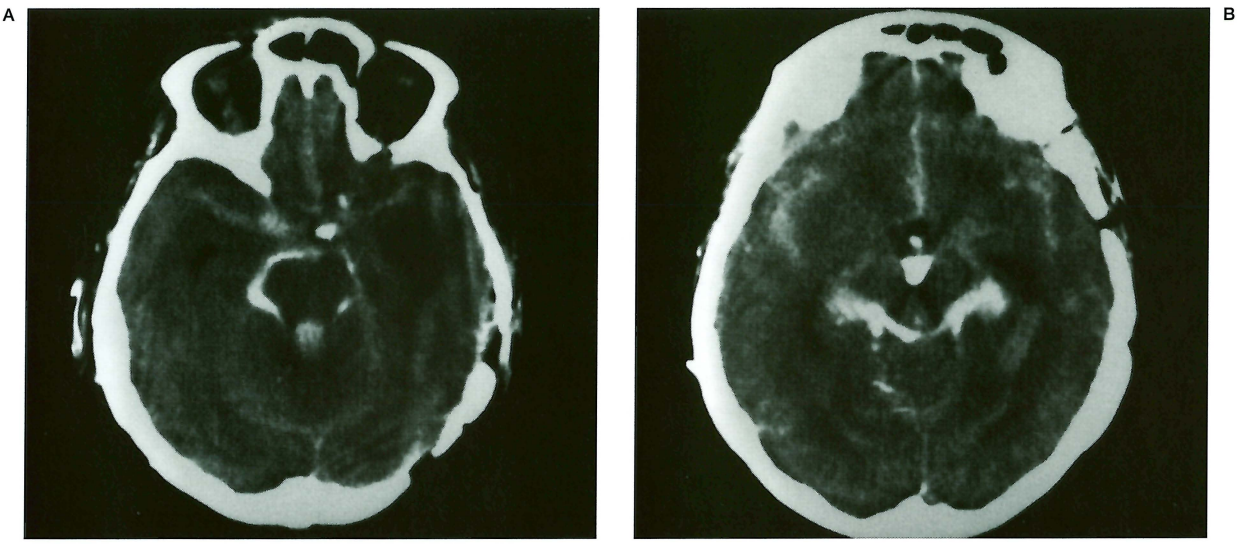

Figure 5 The cranial CT demonstrates the extensive subarachnoid haemorrhage mixed with extravasation of contrast material throughout the basal cisterns (A) and the third ventricle (B).

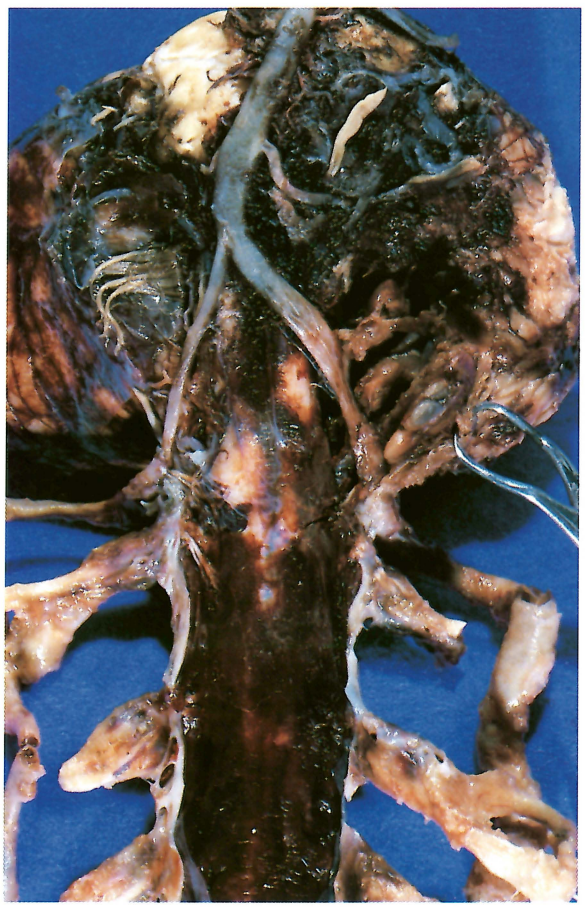

Figure 6 The pathologic specimen shows the extensive haemorrhage in the entire cervical and cranial extracerebral space.

Figure 7 After removal of the blood clots a medium sized aneurysm with the ruptured dome at the angle of the superior cerebellar artery to the BA, is disclosed looking to the right. There is no thrombus or atherosclerotic changes of the parent artery.

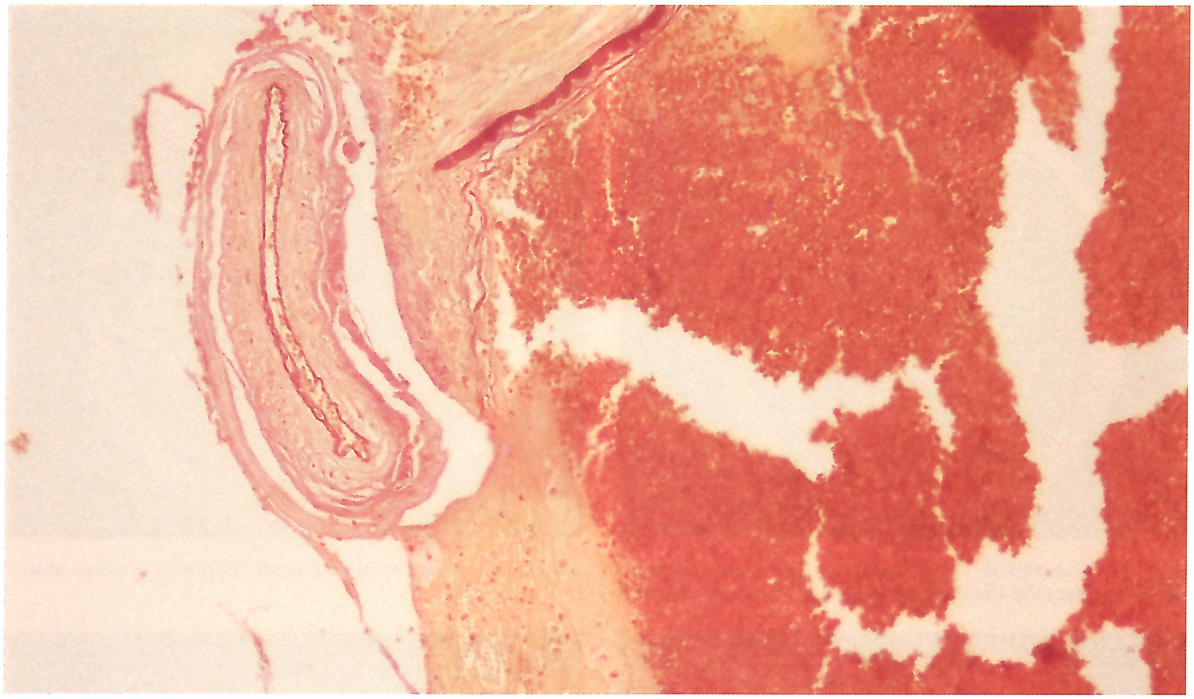

Figure 8 Microscopic finding of the BA at the site of the rupture (at the level of a normal looking pontine branch with normal sized intima and adventitia) discloses an abrupt tear of the internal elastic lamina, a thinning of the adventitial layer and fresh thrombotic material in the vessel lumen. (Formalin fixation, embedding in paraffin, staining of 5 µm thin sections according to the Elasica van Gieson-method, magnification x).

tumour at the skull base and not a suspected associated vascular lesion at the top of the BA and because of the small diameter of the right VA and its consequently low contrast) retrospective analysis of the bi-directional projections of the angiogram did not prove any widening suspect for aneurysm.

In the specimen no findings of atherosclerotic changes such as lipophages and calcifications or cholesterol arose, either in the cerebral or extracerebral vessels. The lack of older thrombotic material or calcification of an aneurysm wall at the site or vicinity of the rupture contradicted the supposition of a pre-existing vascular lesion of a thrombosed, angiographically invisible aneurysm.

The neuropathological changes with different stages of internal elastic lamina leakage are regularly seen in the vessels of patients, who died on a spontaneous rupture of a berry aneurysm of the cerebral vessels [12]. They are also found at the rupture site as in vessels far from the aneurysm, representing and underlining the general pathological changes in the vessel structure in this disease. On the basis of these clinically calm arterial wall lesions, the general blood pressure elevation also led to an elevation of arterial wall stress, resulting in the rupture at the site of the leakage of minimal resistance [13].

Conclusion

The development of cerebral berry aneurysms is thought to be the consequence of an acquired failure of a primarily underlying structural defect of the vessel wall. We suspect these structural defects of the internal elastic membrane and especially of the tunica media to be acquired lesions as well.

Focal rupture of the internal elastic membrane could be induced by mechanical forces and a degeneration of smooth muscle cells in the tunica media could represent an exhaustion of these cells following permanent stimulation (contraction) beyond the physical range. Many trigger-factors are thought to be the relevant. We were able to describe an obviously "spontaneous" development and rupture of a basilar aneurysm within about one hour, when psychic stress is the most likely trigger. As a second result we could prove the existence of vasospasm in acute subarachnoid haemorrhage.

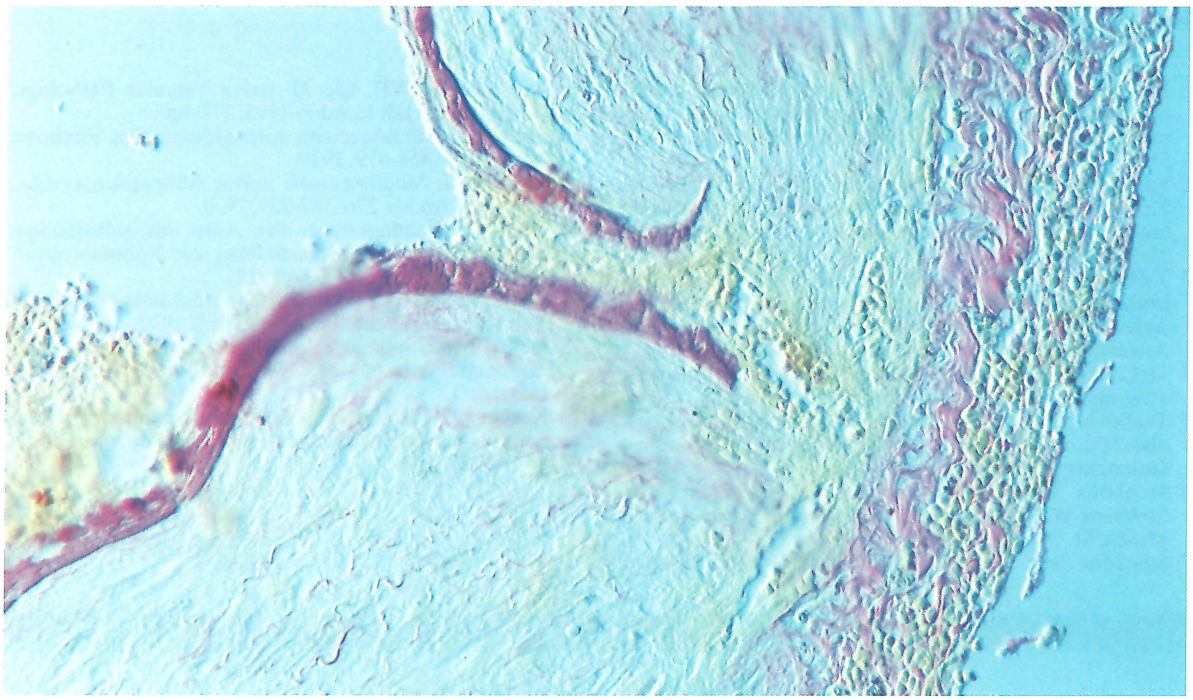

Figure 9 In this microscopic specimen far from the aneurysm another rupture of the internal elastic lamina is seen with a subacute haemorrhage in the adventitial layer, not even reaching the external elastic lamina. (Formalin fixation, embedded in paraffin, staining of 5 µm thin sections according to the Elasica van Gieson-method, magnification x).

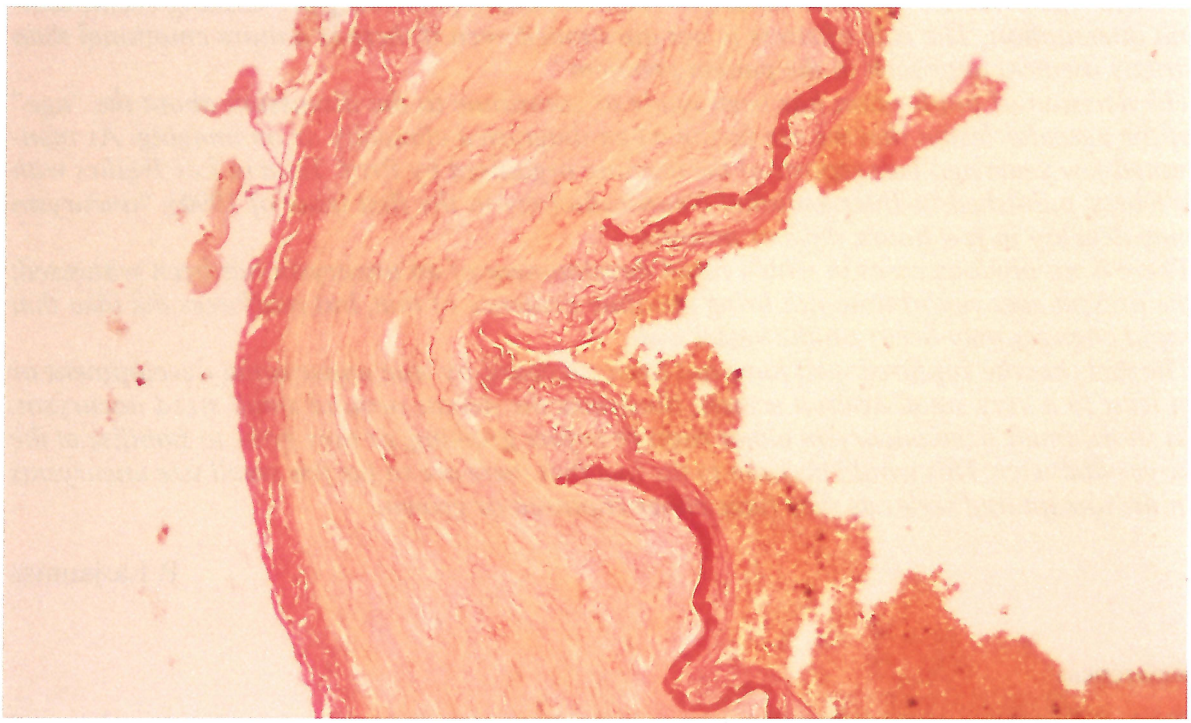

Figure 10 Another microscopic finding at another part of the basilar artery, far from the site of the rupture shows an adventitial scar. Note the tear of the primary internal elastic lamina and the secondary pseudo-lamina, bridging the primary rupture site, where the size of the adventitia is smaller than in the vicinity. (Formalin fixation, embedding in paraffin, staining of 5 µm thin sections according to the Elasica van Gieson-method, magnification x).

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EDITORIAL COMMENT

The paper written by Mrs Müller-Forell has been published despite several criticism made by the review committee. For most of the reviewers, the case is that of "a ruptured artery without true evidence of aneurysmal growth, in a patient presenting pre-existing elastic lamina interruption. The role of stress is not questioned even if there is a more emotional than strictly medical approach to the patient problem".
The decision of publication has been made to contribute to the discussion about the "age" of the vascular lesion that we are facing when diagnosed clinically or on imaging. As mentioned few years ago (see reports of the meeting organised by Prof. Testa in Les Treilles with Wiebers, published in Interventional Neuroradiology in the first issue of 1996), "aneurysm may develop in few hours, days or months, ...".
There is no obvious cases in which rapid development of an aneurysm has been witnessed; the present case report does not bring absolute evidence of that, but reinforces the idea that rapid changes may occur on the vessel wall.
The fact that the ruptured wall have not been preceded by true aneurysmal development or at least to a very small arterial ectasia, illustrates the debate around small sized aneurysm. In short, small sized aneurysm either rupture before growing, or they grow to stabilise at the larger diameter. This would account to the fact that there are so far few small size aneurysms in the unruptured series as they have either ruptured or grown.

P. Lasjaunias

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